



A Case Report of Rare Disease: Adenocarcinoma Esophagus in 12-years-Old Boy in Children's Hospital, Lahore

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ABSTRACT

Introduction: Squamous cell carcinoma of esophagus is more common than adenocarcinoma. It is most common in Asian countries and more prevalent in black population as compared to white population. Its incidence has a decreasing trend, recent years. This disease is very rarely seen in children and adolescents.

Case Report: A 12-years-old boy with moderately differentiated adenocarcinoma of lower esophagus is reported because it is a very rare disease in children. Patient presented with vomiting and dysphagia. Surgery was not possible due to wide spread disease. Chemotherapy followed by radiotherapy was given. Patient started taking semisolids after two courses of chemotherapy.

Discussion: Esophageal carcinoma is disease of old age and most prevalent in Asian countries. Most patients with this disease are male and have age more than 50 years. This disease is very rarely found in children, the youngest patient presented with the disease was eight years old. It frequently metastasizes to lungs, liver and mediastinal lymph nodes.

Key Words: Esophagus, Dysphagia, Adenocarcinoma, Asia, Squamous cell carcinoma, Mediastinal lymph nodes

INTRODUCTION

Patients with esophageal carcinoma present after 5th or 6th decade of life. It is more common in males than in females. Squamous cell carcinoma is more common than adenocarcinoma of esophagus. This disease is more common in Asian countries and more prevalent in black population as compared to white population. Its incidence has a decreasing trend in recent years. This disease is very rarely seen in children and adolescents. Cases reported in younger age group mostly presented with squamous cell carcinoma of esophagus. We present a case of very rare childhood adenocarcinoma of esophagus in a 12-years-old boy.

CASE REPORT

A 12-years-old boy presented with progressive dysphagia for last 6 months and vomiting after every meal for two months. Vomiting was initially with solid food, then progressively with semisolid food, and for two weeks he was unable to take even liquids. His dysphagia was not associated with

any chest or throat pain. He had no history of any corrosive intake. His birth history was insignificant, he achieved all his milestones on time and was vaccinated on time as per Government policy. There was no history of esophageal and gastrointestinal malignancies in 1st or 2nd degree relatives. He lost around 5 kg weight in last 3 months.

Except for low hemoglobin levels (7.8 gm/dl), other lab investigations and chest X-ray were normal. Upper GI endoscopy showed normal hypopharynx, a friable mass seen from 6 to 30 cm from incisors completely obstructing the lumen. Scope could not be negotiated to stomach. Biopsy showed moderately differentiated adenocarcinoma. CT abdomen and chest showed large lobulated circumferential wall thickening involving mid and distal part of esophagus with metastatic pulmonary nodules and calcified hepatic deposits as well as calcified abdominal lymphadenopathy. Figure 1 and 2 shows pulmonary nodules and hepatic involvement.

At time of initial hospital admission, he was unable to take anything orally and it was not possible to pass NG tube due to complete obstruction. Surgical opinion was obtained and

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it was decided that surgery is not possible due to extensive local disease and presence of lung and hepatic metastasis. Feeding jejunostomy was planned but refused by family. It was decided in review board to give systemic chemotherapy followed by radiation. Systemic chemotherapy including paclitaxel and 5-FU given. After two courses of chemotherapy, patient started semisolid diet. Six cycles of chemotherapy completed and radiotherapy was given.



Figure 1: Axial slice of CT scan showed multiple nodules in the pulmonary parenchyma.



Figure 2: Axial slice of CT scan showed multiple nodules in the lung.

DISCUSSION

Esophageal carcinoma is disease of old age. Among the two types of this cancer, squamous cell carcinoma esophagus (SCCE) is more prevalent than adenocarcinoma esophagus (ACE).

A large study analyzed the data of 40 years and concluded that most of the patients with this disease are male and have age more than 50 years. They also stated that although SCC is more prevalent, incidence of ACE is on the rise compared to SCC.¹ Another study stated that esophageal carcinoma is most prevalent in Asian countries and overall incidence is decreasing but cases with ACE are on the rise.² The risk factors for esophageal carcinoma include obesity, gastroesophageal reflux disease, Barrett's esophagus (BE), tobacco use, alcohol and a diet low in vegetables and fruits.³ These patients are old age and mostly men. This disease is very rarely found in children.^{4,5} Only few case reports are found in literature and mostly from south Asia.^{6,7} The youngest patient presented with the disease was eight years old. Almost all of these children presented with squamous cell carcinoma of esophagus. A study conducted in US analyzing the data from 1950 to 2015 found only 3 cases of esophageal cancer below 1 years and only one child has ACE.⁸ Among younger patients risk factors identified for ACE are infrequent teeth cleaning, passive tobacco smoke exposure, and pest infestation of grains.⁹

Our patient was 12 years old boy with ACE, very rarely seen in children. He belonged to sub-urban area and we did not find any risk factor in his history which could be associated to esophageal carcinoma. The clinical presentation of ACE is similar to adults such as weight loss, dysphagia, regurgitation, hematemesis, chest pain and palpable mass whereas our patient presented with weight loss and dysphagia.¹⁰ This cancer frequently metastasizes to lungs, liver and mediastinal lymph nodes and in our patient, it involved lungs, liver and abdominal lymphnodes.¹¹

The management in this situation involves passing jejunostomy or gastrostomy tube to provide nutrition but our patients' family refused so we had to go for total parenteral nutrition. Surgery could not be done due to metastatic disease so he was treated with chemotherapy and radiotherapy. After two courses of chemotherapy, he was able to take semisolid food.

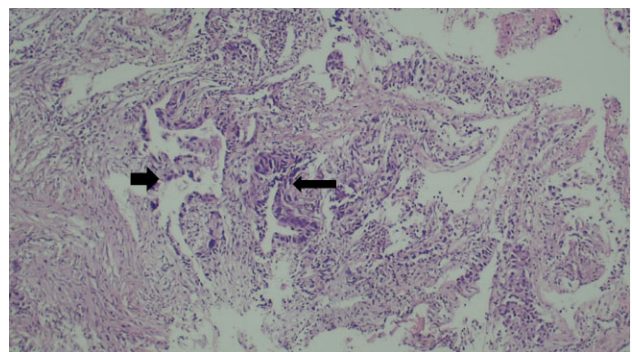


Figure 3: Photomicrograph shows neoplastic cells arranged tubules present in esophageal wall, Moderately Differentiated Adenocarcinoma, pT2 (Arrows) X 20.

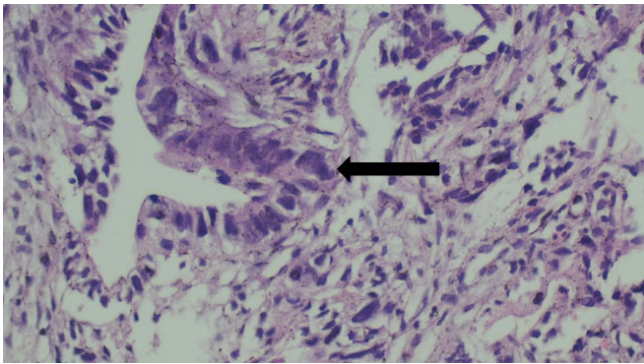


Figure 4: Photomicrograph shows atypical cells with increased N:C ratio lining the glands, Moderately Differentiated Adenocarcinoma, pT2 (Arrows) X40.

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Authors' Contribution:

Dr. Alia Ahmad; Concept of study

Dr. Fariha Sahrish; Planning & Manuscript drafting

Dr. Ayesha Bibi; Study conduction

Dr. Zainab Ehsan; Analysis & interpretation of discussion

Dr. Mahvish Hussain; Facilitation of study and revision for intellectual content

REFERENCES

1. He H, Chen N, Hou Y, Wang Z, Zhang Y, Zhang G, et al. Trends in the incidence and survival of patients with esophageal cancer: a SEER database analysis. *Thoracic cancer*. 2020 May;11(5):1121-8.
2. Xie SH, Lagergren J. Time trends in the incidence of oesophageal cancer in Asia: Variations across populations and histological types. *Cancer Epidemiol*. 2016;44:71-76. doi:10.1016/j.canep.2016.08.002
3. Murphy CC, Claire Yang Y, Shaheen NJ, Hofstetter WL, Sandler RS. An age-period-cohort analysis of obesity and incident esophageal adenocarcinoma among white males. *Dis Esophagus*. 2017;30(3):1-8. doi:10.1111/dote.12526
4. Kumar P, Dana R, Prashant Dadhich, Mehta V, Chaturvedi MK. A Rare Case Report: Carcinoma Esophagus in 12-year-old Girl with Unknown Etiology. *Cancer Ther Oncol Int J*. 2019;14(1):1-3. doi:10.19080/CTOIJ.2019.14.555882
5. Shahi UP, Dattagupta S, Singhal S, Kumar L, Bahadur S, Rath GK. Carcinoma oesophagus in a 14 year old child: report of a case and review of literature. *Tropical gastroenterology: official journal of the Digestive Diseases Foundation*. 1989 Oct 1;10(4):225-8.
6. Allam AR, Fazili FM, Khawaja FI, Sultan A. Esophageal Carcinoma in a 15-Year-Old Girl: A Case Report and Review of the Literature. *Ann Saudi Med*. 2000;20(3-4):261-264. doi:10.5144/0256-4947.2000.261
7. Soni NK, Bikaner PC. Carcinoma of the oesophagus in an eight-year-old child. *The Journal of Laryngology & Otology*. 1980;94(3):327-329. doi:10.1017/S002221510008885X
8. Theilen TM, Chou AJ, Klimstra DS, LaQuaglia MP. Esophageal Adenocarcinoma and Squamous Cell Carcinoma in Children and Adolescents: Report of 3 Cases and Comprehensive Literature Review. *J Pediatr Surg Case Rep*. 2016;5:23-29. doi:10.1016/j.epsc.2015.12.008
9. Buckle GC, Mmbaga EJ, Paciorek A, Akoko L, Deardorff K, Mgisha W, et al. Risk factors associated with early-onset esophageal cancer in Tanzania. *JCO Global Oncology*. 2022 Feb;8:e2100256. doi:10.1200/GO.21.00256
10. Hu N, Wang K, Zhang L, Liu ZJ, Jin Z, Cui RL et al. Epidemiological and clinical features of functional dyspepsia in a region with a high incidence of esophageal cancer in China. *Chin Med J (Engl)*. 2021 Jun 20;134(12):1422-30. doi:10.1097/CM9.0000000000001584
11. Luo LN, He LJ, Gao XY, Huang XX, Shan HB, Luo GY et al. Evaluation of preoperative staging for esophageal squamous cell carcinoma. *World J Gastroenterol*. 2016 Aug 8;22(29):6683-9. doi:10.3748/wjg.v22.i29.6683